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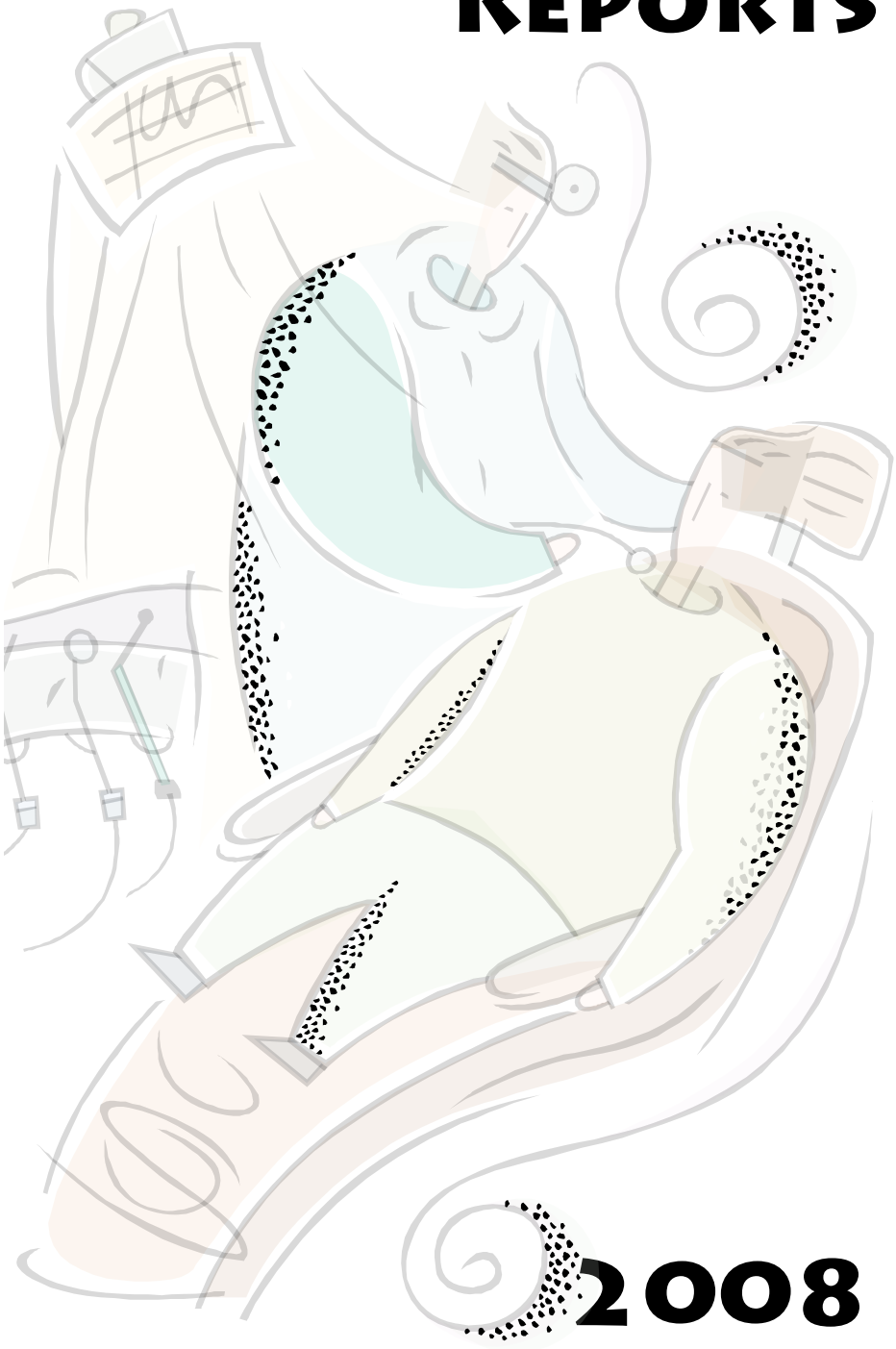

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Oral health status and oral features of Chinese people with Systemic Sclerosis (Scleroderma) in Hong Kong

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Oral health status and oral features of Chinese people with Systemic Sclerosis (Scleroderma) in Hong Kong

Community Health Project
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Abstract

Aims: To study oral health status and oral features of Chinese people with Systemic Sclerosis in Hong Kong.

Materials and method: Chinese people with Systemic Sclerosis attending the Rheumatology Clinic, Queen Mary Hospital were invited to this study. They were informed the study purposes and procedures, and were asked to sign a consent. The study comprised of a questionnaire survey and a clinical examination. In the survey, dental service attendance, oral hygiene habits, and satisfaction with oral health were asked. Xerostomia Inventory was used to investigate subjective dry-mouth symptoms. Salivary function, mouth opening, caries experience, periodontal and oral mucosal status were assessed during clinical examination.

Results: Seventy-seven people were invited and 43 joined this study. Their mean age was 54±12. They practised tooth-brushing daily and about one third had regular dental visit. They all had caries experience with a mean DMFT 11.4. Twenty seven participants (66%) had untreated decay. No one was found periodontally healthy. Most of them (97%) had calculus and the majority (78%) had periodontal pocket. Oral mucosal telangiectasia was common (>80%). Eighteen participants (42%) had reduced mouth opening. Their salivary pH was within normal range. Many experienced subjective dry-mouth symptoms (>70%). Half of them had reduced salivary flow rate, which was found to be associated with dissatisfaction with their dental health ($p=0.05$).

Conclusion: Periodontal pockets and untreated dental caries were common amongst Chinese people with Systemic Sclerosis in Hong Kong. Oral mucosal telangiectasia, reduced salivary flow and reduced mouth opening were also common.

List of Appendices

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Chapter 1 Introduction

1.1 Scleroderma and Systemic Sclerosis

Scleroderma is a symptom of a group of diseases that involve the abnormal growth of connective tissue, which supports the skin and internal organs (NIH, 2007). Originally it was reported by Dr Curzio, an Italian physician, in 1752 describing a young lady whose skin became harden over the entire body (Rodan et al., 1962). According to the American College of Rheumatology (2008), scleroderma has two broad categories which are localized scleroderma (LS) and Systemic Sclerosis (SSc).

LS is a disorder of the skin and sometimes the deeper tissues. The most visible effects of the disease are skin lesions which are often referred as morphea. In some cases, morphea only causes cosmetic problems. However, widespread skin lesions in which scarring spreads down to the underlying structures are called generalized morphea or linear scleroderma.

The second type of scleroderma, SSc, indicates a more extensive skin involvement and even internal organ diseases. SSc is a clinically heterogeneous, systemic disorder which affects the connective tissue of the skin, internal organs and the walls of blood vessels. It is believed that SSc is of autoimmune origin and thus SSc has been grouped under the subject of rheumatology.

1.2 Classification of Systemic Sclerosis

Systemic Sclerosis can be classified into limited Systemic Sclerosis (lSSc), limited cutaneous Systemic Sclerosis (lcSSc) and diffuse cutaneous Systemic Sclerosis (dcSSc) (LeRoy et al., 1988). The most common form of SSc is lSSc (Guern et al., 2004; Scussel-Lonzetti et al., 2002). lSSc and lcSSc often run a fairly benign course due to their relatively minimal skin involvement and late visceral involvement while dcSSc presents with sudden onset and rapid visceral involvement progression (NIH, 2007; Saunders, 2003). Studies showed that early presence (especially within the first year of presentation) of cardiac, pulmonary, gastrointestinal or renal disease was predictive of reduced survival rate (Mayes, 2003). The prognosis and survival rate of dcSSc are worse than other forms of SSc in general.

1.3 Diagnosis of Systemic Sclerosis

Diagnosis of SSc is largely dependent on medical history and physical examination. The signs included changed skin appearance and texture, calcium deposits developing under the skin and changes in the tiny blood vessels at the base of the fingernails. Sometimes haematological screening for Antitopoisomerase-1 or Anti-Scl-70 antibodies (in case of suspected dcSSc) and Anticentromere antibodies (in case of suspected lcSSc) may also be employed to confirm the diagnosis. (NIH, 2007)

Patients suffering from SSc usually exhibit CREST syndrome (Chaffee, 1998). The acrostics CREST stands for Calcinosis (C), Raynaud's phenomenon (R), Esophageal dysfunction (E), Sclerodactyly (S) and Telangiectasia (T). CREST syndrome also has various degree of connective tissue fibrosis. The degree of skin and internal organ involvement are the criteria for the classification of SSc.

1.4 Etiology, pathogenesis, and risk factors of Systemic Sclerosis

The etiology of Systemic Sclerosis remains uncertain despite significant advancement in medicine in recent decades. A commonly accepted cause of SSc is an abnormal immune or inflammatory activity stimulating fibroblasts causing overproduction of collagen (mainly type I and type III) that deposites within connective tissue. It leads to fibrosis of the skin and various internal organs (NIH, 2007).

A study reported the relationship between development of SSc and occupational exposure to epoxy resin, white spirit and aromatic solvents (Magnant et al., 2005). In addition, exposure to cleaning products and solvents is also suspected to be a risk factor for SSc (Maitre et al., 2004).

1.5 Prevalence of Systemic Sclerosis

A summary of epidemiological studies is shown in Table 1. The prevalence rate ranged from 4 to 286 per million and the incidence rate reported varied from 0.6 to 22.8 per million per year. The large discrepancy of the figures may be a reflection of methodological differences in case definition among different regions and countries. However it may also be a true local difference as a result of different susceptibilities to SSc on genetic basis, or differential exposure to putative environmental triggers.

Despite variations of prevalence and incidence rate, a high female to male ratio is a consistent finding. A review study found that lcSSc is more frequently found in European, white American and white Australian groups whereas dcSSc affects more black American, some native American and some Asian populations (Mayes, 2003). Many studies reported a higher incidence at 30 to 50 years of age.

Table 1 Prevalence, incidence and gender ratio of people with Systemic Sclerosis

Investigators (Year)	Study site	Prevalence (per million)	Incidence (per million)	Male : Female Ratio
Robert-Thomas *et al.* (2001)	New South Wales Australia	233	22.8	4:1
Englert *et al.* (1999)	Sydney Australia	86	NA	3:1
Steen *et al.* (1997)	Pennsylvania US	NA	9.6 to 18.7	3:1
Mayes *et al.* (1996)	Detroit Metropolitan US	242	18.7	4:1
Geirsson *et al.* (1994)	Iceland	71	3.8	8:1
Tamaki *et al.* (1991)	Tokyo Japan	38	NA	14:1
Silman *et al.* (1991)	West Midlands UK	31	3.7	6:1
Maricq *et al.* (1989)	South Carolina US	286	NA	NA
Michet *et al.* (1985)	Minnesota US	253	13	NA
Medsger & Masi (1971)	Tennessee US	4 to 28	0.6 to 4.5	NA

1.6 Medical problems associated with Systemic Sclerosis

Coexistence of SSc with other autoimmune conditions is well documented. Simultaneous presentation of SSc with systemic lupus erythematosus and/or polymyositis is termed as mixed connective tissue disorder (Scully and Cawson, 1998). It was found that 14% of people with SSc especially those with lcSSc were complicated with secondary Sjögern's syndrome and was markedly associated with lcSSc (Avouac et al., 2004).

Extent of visceral involvement determines the degree of medical complications (Scully and Cawson, 1998). Dysphagia and reflux oesophagitis are commonly seen in people with SSc having gastrointenstinal involvement (Gonzales and Coleman, 1999; Scully and Cawson, 1998). Pulmonary fibrosis impairs respiration and eventually progresses to pulmonary hypertension in 37% of people with SSc leading to serious cardiac disease (Chang et al., 2006). Vascular involvement causes renal crisis which is an important cause of death (Scully and Cawson, 1998).

1.7 Treatment and therapy of Systemic Sclerosis

At present, there is no effective treatment to control the overproduction of collagen. Treatment and management focus on relieving symptoms and limiting damage (NIH, 2007). Pharmacologically, anti-inflammatory drugs and immunosuppressants are used to tackle the possible underlying rheumatological problems (Lin et al., 2003). Vasodilators, calcium channel blockers and angiotensin converting enzyme inhibitors can be prescribed to control symptoms of visceral complications and Raynaud's phenomenon (Scully and Cawson, 1998). Analgesics may be needed to control joint and muscle pain (NIH, 2007). Non-pharmacological approach such as stretching exercises under the instruction of physiotherapists and occupational therapists is also suggested to prevent loss of joint mobility (NIH, 2007).

1.8 Dental and oral features of people with Systemic Sclerosis

Constriction of the oral orifice is a common finding (Scully & Cowson, 1998). It can cause progressively limited mouth opening which is termed as fish-mouth. People with SSc also have a high prevalence of periodontal disease (Raynaud's & Scleroderma Association, 2008). Clinical findings such as widening of periodontal spaces (Rout et al. 1996; Alexandridis and Stuart, 1984), abnormalities in periodontal microcirculation (Scardina et al., 2005), loss of gingival attachment and prominent buccal mucosal crenations (Eversole et al., 1984) were reported. Other common findings including loss of tongue mobility with fibrotic indurations (Eversole et al., 1984), dry mouth (salivary hypofunction) and telangiectasia (Nagy et al., 1994) were also mentioned in individual reports.

People with SSc are at risk of dental caries, oral ulcers and fungal infections (Raynaud's & Scleroderma Association, 2008). Erosion of facial bones may be found. Individual studies and case reports have shown osseous resorption at mandibular angles, coronoid processes and zygomatic arches (Hopper & Giles, 1982; Rout et al., 1996).

1.9 Studies on dental status of people with Systemic Sclerosis

A Medline search of the literature was performed on May 15th, 2008 and yielded a total of 32 English articles published in the period of 1980 to 2008 bearing keywords of "Systemic Sclerosis" or "scleroderma" and "dental" or "oral features". Most papers were case reports and review articles. Three of them were original articles and 6 additional original articles were identified from cited references of the 32 papers. These 9 original studies, except one in Israel, were conducted on Caucasians in Europe and in North America and were summarized in Table 2. So far there is no study reporting the oral features in Chinese population, this community health project aims to study the oral health status and oral features of Chinese people with SSc.

Table 2 Main findings of clinical studies on people with Systemic Sclerosis

Investigators (Year)	Study site	Sample size	Mean age (range)	Main findings
Poole *et al.* (2005)	Pennsylvania, U S	22 1M,21F	-	Decreased mouth opening Poor oral hygiene
Scardina *et al.* (2005)	Palermo, Italy	15 3M,12F	62±8 (35-72)	Abnormalities in periodontal microcirculation
Scardina (2004)	Palermo, Italy	31 31F	52±12	Trigeminal neuropathy Telangiectasia Retraction of oral frenulum Atrophy of oral mucosa Candidal infection Increased gingival bleeding Decreased mouth opening Salivary hypofunction Increased caries experience Mandibular resorption
Rout *et al.* (1996)	U K	21 6M,15F	54 (36-81)	Widening of periodontal ligament space Tooth resorption Bone resorption
Nagy *et al.* (1994)	Debrecen, Hungary	32 3M,29F	49±11	Telangiectasia of oral mucosa Decreased mouth opening
Wood & Lee (1988)	Canada	31 31F	52±12	Decreased mouth opening Increased caries experience Xerostomia Increased mobility of teeth Thickening of periodontal membrane Erosion of mandible
Alexandridis & Stuart (1984)	Los Angeloes, U S	26	48 (30-85)	Thickening of periodontal ligament space
Eversole *et al.* (1983)	San Francisco, U S	19 2M,17F	52 (25-80)	Symptoms referable to the TMJ Crenation of the buccal mucosa hypomobility of the tongue
Marmary *et al.* (1981)	Jerusalem, Israel	21	46±12 (27-63)	Elevated blood sedimentation rate Elevated antinuclear factor Limited mouth opening Gross jaw bone changes Widening of periodontal ligament space

Chapter 2 Aims and Objectives

Studies on oral health status and oral features of people with Systemic Sclerosis (SSc) were mainly conducted on Caucasians. So far there is no study in English reporting the oral features of Chinese people with SSc. This community health project aims to study the oral health status and oral features of Chinese people with SSc in Hong Kong.

2.1 Aims

The aim of this community health project is to study the oral health status and oral features of Chinese people with Systemic Sclerosis.

2.2 Objectives

The objectives are as follows:

1. To investigate the oral hygiene practice of Chinese people with Systemic Sclerosis in Hong Kong.
2. To study the caries experience of Chinese people with Systemic Sclerosis in Hong Kong.
3. To study the periodontal status of Chinese people with Systemic Sclerosis in Hong Kong
4. To examine the oral mucosal health status of Chinese people with Systemic Sclerosis in Hong Kong.
5. To investigate the severity of subjective dry mouth symptoms in Chinese people with Systemic Sclerosis in Hong Kong.
6. To describe the salivary function of Chinese people with Systemic Sclerosis in Hong Kong.
7. To investigate the satisfaction of oral health amongst Chinese people with Systemic Sclerosis in Hong Kong.

Chapter 3 – Materials and methods

3.1 Study sample

All Chinese people with SSc attending the Rheumatology Clinic, Department of Medicine, Queen Mary Hospital were invited to participate in this study by telephone. The study was carried out in the Prince Philip Dental Hospital (PPDH) from the 25th to 29th February, 2008. The progress note of this community health project can be found in Appendix 1.

3.2 Pilot study

The study comprised of two parts. The first part was a questionnaire survey. The second part was a clinical assessment which consisted of a salivary function test and an oral examination. Before the main study, a pilot study was carried out on three middle aged normal individuals to let the group familiarize with the rundown and to estimate the time required for the whole procedure. In this pilot study, the questionnaires were tested to ensure a lay person could understand the wordings. The precision of the measurement of the volume of saliva collected was reviewed. The caries and periodontal status assessment as well as maximum mouth opening measurement were also evaluated. An overall evaluation was conducted to revise the rundown and the procedures before the main study.

3.3 Registration

All participants were informed of the study purposes and procedures by an interviewer. Then, they were asked to sign written consents (Appendix 2, 3 and 4). After that, they were registered as patients for teaching and research purposes in PPDH. The medical history of the participants was checked before commencement of the study (Appendix 5).

3.4 Questionnaire survey

The questionnaire survey was the first part of the study. In this survey, the participant's personal data, dental service utilization, oral hygiene habits, smoking and alcohol drinking habit, perceived dental health and satisfaction with oral health were asked (Appendix 6 and 7).

The Xerostomia Inventory (XI) (Thomson et al., 1999) was used to investigate the severity of subjective dry-mouth symptoms. It contained 11 questions on participants' perception on dryness of the nose, mouth and skin of the face, difficulties encountered in swallowing and eating food, etc. These questions were assessed by patients using a 5-point Likert scale (Appendix 8). Scores of 1 to 5 were given to never (1), hardly ever (2), occasionally (3), fairly often (4) and very often (5).

The scores of the 11 questions were summated to give a single XI score, ranging from 11 to 55. A higher XI score indicates more severe subjective symptoms of xerostomia. Data collected from individual question were reduced to a dichotomy. The responses 'fairly often' and 'very often' indicated a negative impact. The percentage of the study groups with negative impact can be calculated for each individual question.

For participants who had reading difficulties and were illiterate, questions from the questionnaires were asked and explained by a trained interviewer. Completed questionnaire forms were checked independently by another interviewer to ensure there were no missing or unclear answers.

3.5. Salivary function test

After the questionnaire survey, the participants' salivary function was assessed. Those who were suffering from sialadenitis were excluded. Salivary flow rate and acidity of saliva are measured as an attempt to reflect the salivary function test this group of patients. The participants were asked not to eat, drink and smoke for an hour before the sialometric assessments.

The participants were asked to rinse their mouth with 10ml phosphate-buffered saline for 30 seconds. They were then asked to expectorate their saliva every minute into a disposable specimen bottle for 10 minutes. This procedure was repeated to study the stimulated whole salivary flow rate by asking the participants to chew on a piece of 15mm sterilized silicone rubber tubing (Masterflex 964DD-14, Cole-Parmer, Illinois, USA) for 5 minutes. They were then asked to expectorate all saliva stimulated into another disposable specimen bottle.

The resting and stimulated salivary flow rates expressed as ml/min were determined following the guidelines described by Navazesh and Christensen (1982). The salivary pH was measured with a pH meter (Sentron 501Pocket FET pH meter, Sentron, Washington, USA). All measurements were made at the chairside immediately after saliva collection.

3.6 Clinical assessment

The clinical assessments which included an extra-oral and an intra-oral examination were performed according to the guidelines provided by World Health Organization (WHO, 1997) (Appendix 9). In addition, measurement was made to assess maximum mouth opening. The assessments were performed by two calibrated examiners using ruler, dental mirror, straight probe and CPI probe. Cotton roll and gauze were used if necessary for moisture control and removal of debris on the tooth surfaces.

Calibration exercises were performed at the Reception and Primary Care Clinic, PPDH on 10 out-patients to ensure a good agreement between the two examiners in the assessment. In the main study, about 10% of the participants were re-examined to assess the inter-examiner agreement.

The assessment started with extra-oral examination. The TMJ was assessed for any signs and symptoms of TMJ disorders such as clicking sound during mouth opening and closing and tenderness of the joint area on palpation.

The maximum mouth opening was measured by the inter-incisal distance plus the overbite (Wood and Branco, 1979). The overbite was obtained with teeth in centric occlusion. The inter-incisal distance was measured by the distance between maxillary and mandibular central incisors in the midline when the mouth was maximally opened. This measurement of inter-incisal distance was taken three times in succession. Participants with median diastema more than 5mm or missing central incisors were excluded from this part of investigation.

The intra-oral examination evaluated the oral soft tissues and teeth. Oral mucosa condition was examined under optimal lighting using the WHO criteria (WHO, 1997) (Table 3).

Table 3 Codes used in oral mucosa condition assessment

Code	Criteria
0	No abnormal condition
1	Malignant tumour (oral cancer)
2	Leukoplakia
3	Lichen planus
4	Ulceration(aphthous, herpetic, traumatic)
5	Acute necrotizing gingivitis
6	Candidiasis
7	Abscess
8	Other condition (specify if possible)
9	Not recorded

Oral mucosal telangiectasia and tongue-tie were recorded as present (code 1) or absent (code 0) separately. Diascopy test was used to examine blanching of telangiectasia lesions. The wearing of denture was also recorded as present (code 1) or absent (code 0) in the upper and lower arches separately.

Community periodontal index (CPI) was used to assess the periodontal condition. The mouth was divided into six sextants which were defined by FDI tooth numbers 18-14; 13-23; 24-28; 38-34; 33-43; 44-48. Six index teeth (all first molars, upper right central and lower left central incisors) were examined. If no index tooth was present in a sextant qualifying for examination, all the remaining teeth in that sextants were examined and the highest score was recorded as the score for the sextant. In this case, distal surfaces of the third molars were not scored. The recording codes are shown in Table 4.

Table 4 Codes used in periodontal assessment

Code	Details
0	Healthy
1	Bleeding observed, directly or by using a mouth mirror, after probing
2	Calculus detected during probing, all black band on probe visible
3	Pocket 4-5mm, gingival margin within black band on the probe
4	Pocket >6mm, black band on the probe not visible
X	Excluded sextant (< 2 teeth present)
9	Not recorded

The caries status was assessed according to the criteria suggested by WHO (1997). The caries experience was measured with DMF index. Status of each tooth was recorded with codes shown in Table 5.

Table 5 Codes used in caries assessment

Code	Tooth status
0	Sound
1	Decayed
2	Filled, with decay
3	Filled, without decay
4	Missing, as a result of caries
5	Missing, any other reason
6	Fissure sealant
7	Bridge abutment, crown, veneer, implant
9	Not recorded

3.7 Report of assessment and follow-up

Each participant was informed of the results after the assessment. An assessment report (Appendix 10 and 11) and a souvenir were given. A follow-up appointment for scaling and oral hygiene instruction by dental hygienist and a review appointment with a dentist were given. Those participants with caries were advised to seek care from private dentists or be assessed for suitability for treatment by dental students as teaching cases in PPDH.

3.8 Data entry and statistical analysis

The data from the questionnaires and charting sheets were entered into Microsoft Excel 2007 and proof-read by two members separately. Inter-examiner reproducibility was computed using Cohen's Kappa statistics. The data were analyzed using SPSS 15.0 (SPSS Inc., Chicago, USA) for descriptive frequencies.

Student t-test was used to study the dental visit behaviour, salivary flow rate and reduced mouth opening on the caries experience of the participants. Chi-square test was used to study the dental visit behaviour, dental satisfaction, salivary flow rate, reduced mouth opening on the need for advanced periodontal therapy of the participants. Analysis of variances (ANOVA) was used to study the caries experience of the participants and severity of subjective dry-mouth symptoms on dental satisfaction. The cut-off point for statistical significance was set at 0.05.

Chapter 4 Results

4.1 Sample size

All 77 people (4 male, 73 female) with scleroderma registered in the Rheumatology Clinic were invited by telephone for this study. A total of 43 individuals, 1 male and 42 female, participated in this study. The response rate was 56%. Hospitalized and institutionalized patients were not able to participate in this study due to lack of human resources to escort them. The age of the participants ranged from 29 to 79 and their mean age was 54±12. Two participants were edentulous.

4.2 Past dental history

There were 15 participants (35%) who visited dentists within 1 year preceding to this study. 11 participants (26%) had visited dentists one to three years ago and 17 participants (39%) had not visited any dentists for more than 3 years. In this study, the 15 participants who had seek dental consultation within 1 year preceding this survey are considered as having regular dental visit while the remaining are considered to be not having regular dental visit. The sample group is reduced to a dichotomy for the data analysis.

4.3 Oral hygiene habits

All dentate participants (41) practised daily tooth brushing. Most of them (36, 88%) brushed at least twice daily. Other cleaning aids, including toothpick, mouthrinse, inter-dental brush, dental floss were all commonly used by the participants. Many of them (25, 61%) used toothpicks daily while 13 participants (32%) used dental floss or inter-dental brush daily. About one-third of them (13, 32%) had mouth-rinsing habit.

4.4 Smoking and alcohol drinking habits

Only 1 participant reported smoking for 10 years while others had no smoking habit. None of them reported habit of alcohol drinking.

4.5 Salivary function

One participant considered spitting of saliva continuously for ten minutes was disgusting and refused to perform the salivary function assessment. Among the remaining 42 participants, the mean resting salivary flow rate was 0.18±0.20ml/min which was lower than the suggested normal range (0.3-0.4ml/min), but the difference is not statistically significant. The majority of the participants (35, 83%) had resting salivary flow rate below the normal range; and only 1 participant (2%) was found to be above.

The participants' mean stimulated whole salivary flow rate was 0.6±0.5 ml/min. Half of the participants (21, 50%) had stimulated whole salivary flow rate below the suggested normal range (0.5-1.0 ml/min) and 7 participants (17%) were found to be above.

In this study, 21 (50%) participants were classified as having a reduced salivary flow rate when both the resting and stimulated salivary flow rates were lower than the normal range.

None of the participants had unstimulated salivary pH less than the normal range (pH= 6.0-6.5). But most participants (n=39, 93%) had unstimulated salivary pH higher than the normal range with a mean of 6.9±0.4. No participant had stimulated salivary pH lower than the normal range (pH = 6.0-7.0) and there were 12 participants (29%) who had stimulated salivary pH higher than the normal range. The mean of the participants was 7.1± 0.4.

4.6 Subjective symptoms of dry mouth

The mean score of on the severity of subjective dry mouth symptoms of the 43 participants using xerostomia inventory was 25.2± 9.9. 31 participants (71%) had a sum of score below 33 and experienced subjective dry-mouth symptoms.

The number of participants with negative impact on the specific symptoms is shown in Table 6. The most common symptom noted by participants was “the skin of my face feels dry” (18, 44%).

Table 6 Negative impacts on items of xerostomia inventory

Item of Xerostomia Inventory	No. of people (%) with negative impacts
The skin of my face feels dry	18 (44%)
My eyes feel dry	12 (28%)
My mouth feels dry	12 (28%)
My lips feel dry	12 (28%)
I have difficulty in eating dry foods	10 (23%)
I get up at night to drink	7 (16%)
My mouth feels dry when eating a meal	7 (16%)
I have difficulties swallowing certain foods	4 (8%)
I suck sweets or cough lollies to relieve dry mouth	4 (9%)
I sip liquids to aid in swallowing food	4 (9%)
The inside of my nose feels dry	3 (7%)

4.7 Mouth opening

Two edentulous participants and 7 people who had at least one reference central incisor missing were excluded in this measurement. For the remaining 34 participants, the mean maximum inter-incisal mouth opening was 40±7mm. It ranged from 30 to 51 mm. An inter-incisal distance less than 40mm was considered less than normal and 18 participants (42%) had reduced inter-incisal distance.

4.8 Temporomandibular joint assessment

Eight participants (19%) had clicking on mouth opening and 3 (7%) had tenderness upon palpation on TMJ.

4.9 Oral mucosal health status

Oral mucosal telangiectasia was observed in 35 participants (81%) and was commonly found on the lateral border of the tongue and buccal mucosa. No clinical evidence of common oral mucosal lesions such as candidal infection, recurrent aphthous ulceration and lichen planus was found.

4.10 Inter-examiner agreement

Two calibrated examiners were involved in assessing the caries status and the periodontal status in the study. Duplication examination was carried out in 5 (12%) participants. The Kappa statistics in the assessment of caries and periodontal status were 0.98 and 0.95 respectively.

4.11 Caries experience

The caries experience of the participants was reflected by calculating the mean DMFT scores. The mean DMFT was 11.4±8.8. The decayed (DT), missing (MT) and filled (FT) teeth were 2.0±2.3, 5.3±8.0 and 4.1±4.2 respectively. They contributed 18% (DT), 46% (MT) and 36% (FT) to the caries experience respectively. No participant was found with no caries experience (DMFT=0) and 27 participants (66%) who had at least one untreated caries were found. They had an average of 3.1 decay teeth. There was no association found between caries experience of SSc people with dental visit behaviour, salivary flow rate or extent of mouth opening (Table 7).

Table 7 Caries experience and dental visit behaviour, salivary flow rate and extend of mouth opening of people with Systemic Sclerosis (N = 43*)

Caries experience	Regular Dental Visit Behaviour		Reduced Salivary Flow Rate		Reduced Mouth Opening*	
(DMFT)	Yes	No	Yes	No	Yes	No
No. of people	15	28	22	21	18	16
Mean DMFT± SD	11.6± 8.5	11.3± 9.1	9.2± 7.0	13.5± 10.0	11.1± 5.9	8.4± 7.6
P value	0.92		0.11		0.28	

* N=34 for assessment of reduced mouth opening

4.12 Periodontal status

Two edentulous participants were excluded from the periodontal status examination. None of the participants was found to have healthy gum, i.e. scored CPI=0 as the highest score among the 6 sextants examined (Table 8). Most of the participants (98%) had calculus and many (78%) had periodontally involved teeth (scored 3 or 4).

Table 8 Periodontal status of people with Systemic Sclerosis (N=41)

Participants with	%	Sextant score count	%
Highest CPI = 0	0	Healthy (CPI = 0)	0
Highest CPI = 1	2	Bleeding only (CPI = 1)	14
Highest CPI = 2	20	Calculus (CPI = 2)	45
Highest CPI = 3	40	Pockets (CPI =3 or 4)	33
Highest CPI = 4	38	Excluded	8
Total	100	Total	100

There was no statistically significant difference between the presence of periodontal pocket ≥4mm for SSc people with their dental visit behaviour, salivary flow rate and extent of mouth opening (Table 9).

Table 9 Need of advanced periodontal treatment and dental visit behaviour, salivary flow rate and extent of mouth opening of people with Systemic Sclerosis (N = 41*)

Need of advanced periodontal treatment (CPI = 3 or 4)	Regular Dental Visit Behaviour		Reduced Salivary Flow Rate		Reduced Mouth Opening*	
	Yes	No	Yes	No	Yes	No
Yes (31, 76%)	9	22	15	16	13	12
No (10, 24%)	6	4	5	5	5	4
P value	0.13		0.93		1.00	

* N=34 for assessment of reduced mouth opening

4.13 Perceived needs

Only perceived dental needs of the dentate participants were investigated. 18 participants (42%) thought they had existing caries in which 16 of them (89%) were diagnosed to have active caries in the clinical examination. Among the remaining 23 participants (59%) who either did not know or thought they did not have any caries, 11 of them (48%) were diagnosed to have active caries. There was a statistically significant association between the perceived needs for treatment and the presence of active caries (p=0.01).

Nine participants (22%) thought they had existing periodontal health problem in which 8 of them (89%) were found to have periodontal pocket. For the remaining 32 participants who were uncertain or thought they did not have any periodontal health problem, 23 of them (72%) were found to have periodontal pocket. There was no association between the perceived needs for periodontal treatment and presence of periodontal pockets ≥4 mm. (p=0.22).

4.14 Past dental history and attitudes towards dental treatments

There were 15 participants (35%) who visited dentists within 1 year preceding this survey, in which 10 of them (67%) were diagnosed to have active caries and 9 of them (60%) were diagnosed to have periodontal pocket. Eleven participants (27%) had visited dentists within one to three years and 7 of them (64%) were diagnosed to have active caries while 9 of them (82%) were found to have periodontal pocket. There were 15 participants (37%) had never visited or visited dentists more than 3 years preceding to this survey. Amongst them 10, (67%) were diagnosed to have active caries and 13 (87%) were found to have periodontal pocket.

For those 20 participants who didn't seek dental treatment despite of the fact that they thought they had dental diseases, their reasons for not seeking dental treatment included high dental fee (n=9, 45%), dental phobia (n=7, 35%), no time to see a dentist (n=5, 25%), afraid of dental pain (n=4, 20%) and difficulty in accessing to dental clinics (n=2, 10%) while 5 of them(25%) put in other reasons such as difficulty in opening their mouth or old age, etc.

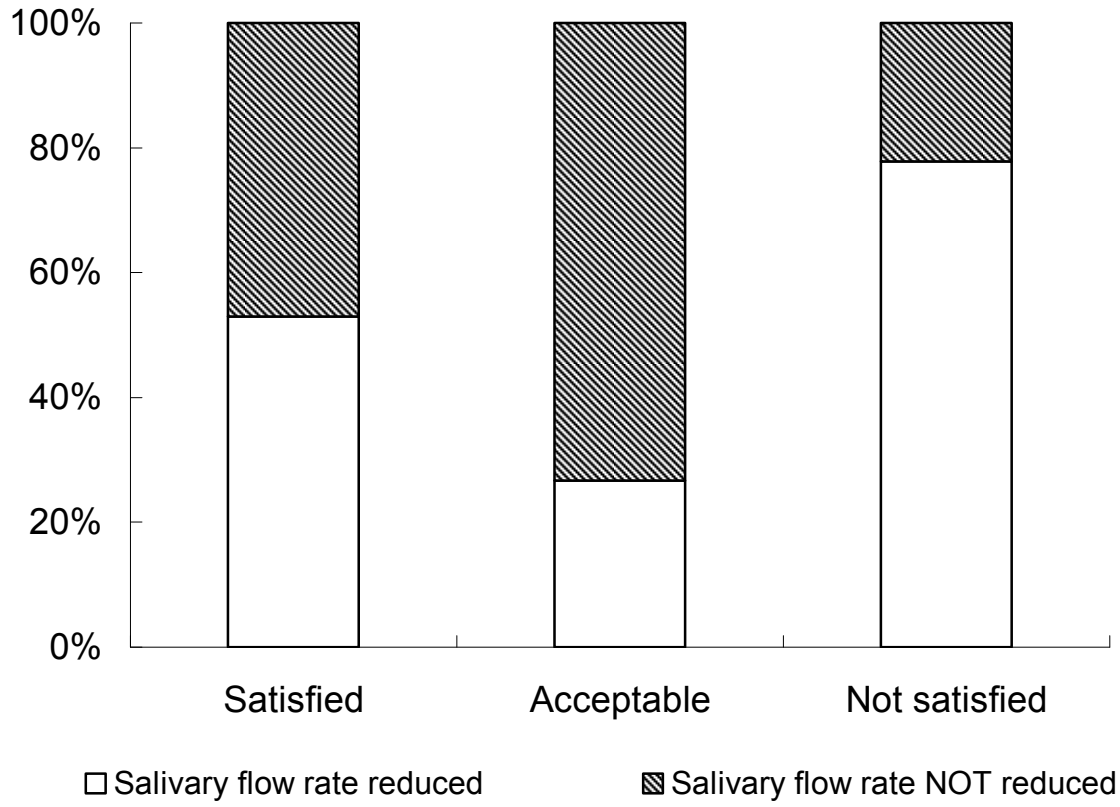
4.15 Satisfaction with oral health

The number of participants and their satisfaction with their oral health is shown in Table 10. There was no significant relationship between their satisfaction of oral health and their caries experience and the presence of advanced periodontal disease. Satisfaction of oral health also had no significant association with their subjective dry mouth symptoms in this study. However, the results indicated that reduced salivary flow rate had an adverse effect on the satisfaction of oral health (p=0.05) (Figure 1).

Table 10 Satisfaction of oral health of people with Systemic Sclerosis and their caries experience, presence of advanced periodontal disease and salivary flow rate (N = 41)

Satisfaction with oral health (n,%)	Caries experience (DMFT)	Advanced Periodontal Disease (CPI ≥ 3)		Reduced Salivary Flow Rate		Subjective Dry-mouth Symptoms (XI score)
	Mean ± SD	Yes	No	Yes	No	Mean ± SD
Satisfied (17,41%)	9.2±6.3	15	2	9	8	25.2±10.3
Acceptable (15, 37%)	10.5±9.2	9	6	4	11	25.9± 10.5
Not satisfied (9,22%)	12.4±8.0	7	2	7	2	22.7±9.6
P value	0.61	0.18		0.05		0.74

Figure 1 Oral health satisfaction and salivary flow rate



Chapter 5 Discussion

5.1 Study sample

It is considered many people with SSc in Hong Kong would be referred to the Rheumatology Clinic of the Queen Mary Hospital for medical care. Within the confined resources and time available, the study managed to examine all 43 patients that accepted our invitation. The response rate of our study was considered satisfactory.

In the nine reports found in the literature search in Chapter 1(Table 1), many of them had sample sizes between 20 and 30. The one with the largest sample size was 32. Despite a larger sample was recruited in this study, the studied participants were by no means a representative sample. Hospitalized and institutionalized people with SSc might have had physical difficulties which hindered them to join this study. However, this study provides useful information of oral health status of Chinese people with SSc. It allows a rational understanding of the oral health status of people with SSc in Hong Kong.

There is no statistics available for the prevalence or incidence of people with SSc in Hong Kong. It is estimated that the prevalence of SSc amongst Chinese in Hong Kong is about 70-100 per million, compared with 31-253 reported in the literatures. We found most people affected were female and only one male was affected among the 43 participants. This finding is similar to other studies (Table 1).

5.2 Questionnaire survey and clinical assessment

Both the clinical examination and the questionnaire survey were conducted smoothly and efficiently. This could be attributed to the pilot study which allowed an evaluation of trial run before the main study. The questionnaires were found to be easily understood. The close-ended questions were quick to complete. No missing or unclear answer was found because each questionnaire was double checked by another interviewer independently so that clarification or amendment could be made immediately before the participant left.

Two examiners were involved in clinical assessment of the caries and the periodontal status in this study. The level of inter-examiner agreement was good compared with previous epidemiological studies (Faculty of Dentistry, 2006 and 2007). Well-defined diagnostic criteria, calibration exercise and the pilot study all contributed to the good level of inter-examiner agreement in the main study.

The pH meter used and the method of measurement suggested by Navazesh and Christensen (1982) allowed an assessment with good precision on salivary pH and flow rate. In the Chinese culture, it is impolite and childish to play with saliva. It is understandable that the salivary function test might not be accepted by some Chinese participants. A clear explanation with patience on the significance and importance of the salivary function test was necessary for good cooperation of the participants.

5.3 Salivary function

The salivary pH value of more than one third of people with SSc was higher than normal. The salivary flow rate was found to be reduced in almost half of the people with SSc and this may give rise to an increased risk of caries, problems of denture, altered taste sensation and pathological conditions such as burning mouth syndrome. Temporary oral dryness may be caused by drugs, dehydration and stress etc. A longitudinal assessment is necessary to monitor the xerostomic condition of people with SSc because salivary hypo-function is a part of disease symptoms of scleroderma (Rasker, 1990; Wood, 1988).

The decreased salivary flow rate may partly due to fibrotic processes in salivary glands. Although there is no universally determined critical value below which salivary flow is considered pathological, Bertram (1967) proposed whole resting saliva rate less than 0.1ml/ min was regarded as a sign of salivary hypo-function. People with SSc should also be checked for Sjögren's syndrome as studies reported a 17% to 29% prevalence of Sjögren's syndrome in scleroderma patients (Osial, 1983; Drosos, 1988; Cipoletti, 1977).

It would be desirable to check the buffering capacity of the saliva of the people with SSc to investigate if it is affected by SSc. CRT buffer caries risk test (CRP® buffer, Vivadent, Liechtenstein, Germany) could be employed, but it was not available during the study period.

5.4 Subjective symptoms of dry mouth

Xerostomia Inventory (XI) was used to measured subjective symptoms of dry-mouth (Thomson, Chalmers et al., 1999). The XI was used to measure the severity in xerostomia during treatment of dry-mouth in Sjogren's syndrome (McMillan, Leung et al., 2004). In this study, mean XI score was used to compare the adverse effect of dry-mouth with non-xerostomic people of similar age (Thomson and Williams, 2000 and Thomson, 2007). It was found that the mean score for people with SSc was only slightly higher. Therefore, SSc did not have a significant impact on the subjective symptoms of dry-mouth in this study.

Subjective feeling of dry-mouth was reported in about a quarter of the participants with SSc, whereas the prevalence of xerostomia in scleroderma patients reported in previous studies was 42 to 70% (Eversole et al.,1984; Wood and Lee, 1988).

The prevalence of xerostomia in our study group was significantly lower than the percentage of subjects reporting negative impact for general dry mouth in nasopharyngeal carcinoma survivors and in Sjogren's syndrome patients (McMillan et al., 2004a; McMillan et al., 2004b).

About one third of the people with SSc in this study showed a XI score equal to or more than 30. This could mean that SSc has a larger effect on some Chinese people with SSc in Hing Kong or more severe self-reported symptoms of dry mouth could be resulted from more severe scleroderma. Further clinical studies of these patients can be done to provide more information about possible correlation between SSc and oral health-related quality of life (OHRQOL).

5.5 Mouth opening

An established maxillo-facial clinic in Zurich accepted a 38 mm incisal edge distance as the minimum limit for adults (Landtwing, 1978). The appropriateness of applying such a guideline on participants in this study is uncertain due to different ethnicity. A report suggested a maximum incisal opening of 40mm or less would appear to warrant investigation as to its etiology (Sheppard and Sheppard, 1965). In this study, an inter-incisal distance less than 40mm was considered less than normal and almost half shown reduced inter-incisal mouth opening. A study reported that maximum mouth opening diminishes with age (Mezitis et al., 1989).

Apart from age, pathology and injuries leading to TMJ pain in the maxillofacial region can influence mouth opening. In this study, TMJ pain was not common. However, almost half of the people have noticeable reduced mouth opening. This is probably due to pathological changes in connective tissue resulting in a constricted oral orifice.

5.6 Mucosal changes

Oral mucosal telangiectasia was prevalent amongst the participants of this study. The common sites were the lateral border of the tongue and buccal mucosa of cheek. This finding agrees with two previous reports (Nagy et al., 1994; Scardina, 2004). It was also reported that telangiectasia was commonly seen on the face, lips, tongue and fingers (Chaffee, 1998). Telangiectasia manifests as small red spots by swelling of tiny blood vessels beneath the skin. It could cause cosmetic problems if appears on the hands and faces (NIH 2007).

Previous studies found this group of patients had restricted tongue mobility (Naylor, 1982; Eversole et al., 1984; Wardrop and Heggie, 1987). Fibrotic changes of the tongue

or lingual frenum may attribute to this phenomenon. In this study, it was observed that some participants were having hypo-mobility of the tongue. Despite some mucosal lesions reported amongst people with SSc in some reports (Scardina, 2004, Raynaud's & Scleroderma Association, 2008), no common oral mucosal lesions such as candidal infection, recurrent aphthous ulceration or lichen planus were observed in this study.

5.7 Caries status

The oral hygiene practice of Chinese people with SSc in Hong Kong was generally good as almost all brushed their teeth at least twice a day. However this study found that only a low percentage of people with SSc had regular dental visit to dentists. Compared to the caries experience of the general population in Hong Kong (Department of Health, 2002), a similar caries experience among people with SSc was observed. However, they had more untreated decay (DT) when compared with those of Hong Kong adults.

This study found an average of two untreated caries in Chinese people with Systemic Sclerosis. Literature reported people with SSc had oral features that may complicate dental treatment. It is therefore essential for these people to receive regular dental service for good oral health. It is imperative that they should be educated the importance of regular dental visit for optimal oral health and prevention of oral diseases.

This study found that participants with dental caries knew they need dental treatment. It is because there is a significant association between their perceived needs and the presence of untreated caries.

Reasons for not seeing dentists of the 20 participants who have untreated decayed teeth included dental phobia, high cost of dental care and too busy for treatment. There are dental services run by non-profit making organizations in Hong Kong which provide dental services at a reasonable cost and those families from low social class may be able to receive financial assistance from the government. These services should be informed to the needed patients. As neglecting oral diseases will worsen the outcome of dental care and incur a higher treatment cost in the future, oral health education should also be promoted to people with SSc for earlier treatment and prevention of dental diseases.

5.8 Periodontal status

This study reflected that periodontal disease was very prevalent and severe among Chinese people with SSc in Hong Kong. None of our participants had healthy periodontium, and almost all of them (98%) had calculus deposits. The majority of them (78%) had periodontal pockets and needed advanced periodontal care. Their periodontal condition was worse than that of the general population (calculus 43%; periodontal pockets 55%) in Hong Kong (Department of Health, 2002).

A clinical study found the ability to open the mouth as shown by the oral and incisor aperture was significantly decreased in patients with poor oral hygiene (Poole et al., 2005). This study thus concluded that they had a higher level of periodontal disease. Some also suggested the poor periodontal health may be related to reduced vascularity with resulting tissue ischemia in scleroderma individuals (Wood et al., 1988). However, this study did not find a significant effect of reduced mouth opening on the severity of periodontal disease.

5.9 Dental management of people with Systemic Sclerosis

Caries and periodontal disease prevention are the prime concerns in the treatment plan for SSc patients. It is because the protection from saliva is commonly impeded by salivary hypo-function. Their oral hygiene is also compromised due to impaired hand dexterity of oral hygiene tools handling (Poole et al., 2005). Child-sized soft bristle tooth brushes with wide flat handles are suggested for SSc patients (Poole et al., 2005). Fluoride mouthrinse (without alcohol) and chlorohexidine mouthrinse can be prescribed as an adjunct preventive measure. Preventive care is very important as these patients are more difficult to handle for restorative care.

Limited mouth opening makes dental treatment difficult and sometimes impracticable (Scully and Cawson, 1998). Studies suggested stretching exercises by placing the thumbs in opposite corners of the mouth and pulling outwards. In addition, oral augmentation using tongue depressors between the back molars may increase oral aperture (Poole et al., 2005; Naylor et al., 1984; Pizzo et al., 2003). The use of paediatric intra-oral films, head lamps, miniature dental handpiece with drill with small head and triple-angled instruments may be used to facilitate dental operations (Poole et al., 2005).

Treatment options for people with SSc are often limited as dental implants and fixed prostheses are generally not recommended. It is because people with SSc often have sclerodactyly. Their oral and prosthetic hygiene maintenance is often compromised by their inability to floss. In cases with advanced SSc, removable partial denture (RPD) seems to be the only possible treatment option.

Prosthetic management is difficult as limited access for insertion of impression tray and prosthesis resulting from reduction in elasticity of the mucosal membrane. Moreover, normal anatomical landmarks could not be expected due to soft tissue atrophy and hard tissue resorption (Samet et al., 2007).

Sectional impression technique and specially designed bi-part hinge tray controlled by orthodontic elastic band and dental floss were advocated in a case report for impression taking (Yenisey et al., 2005). Collapsed mandibular RPD with lingual midline hinge could be used (Yenisey et al., 2005). Another report suggested the use of flexible Valplast RPD for SSc patients (Samet et al., 2007).

Dry mouth (Xerostomia) is common among people with SSc and affected half of the participants in this study. Moreover, this study also found reduced salivary flow affecting the satisfaction of oral health. Dry mouth is a common cause of halitosis. Reduction in salivary flow will shift the balance of the oral microflora and hence contributes to the dominance of some bacteria in oral plaque causing diseases. Some studies suggested daily use of sugar-free chewing gum to increase the salivary flow rate (Pankhurst et al., 1996).

Chemical salivary stimulants have not gained wide popularity because they are not very effective and associated with inherent adverse reactions. Trials with prilocapine in SSc patients significantly increased submandibular and parotid gland secretions over baseline levels for only 1 to 2 hours after administration, but not after 4 hours (Fox et al., 1991).

Many participants resorted to the use of traditional Chinese medicine for long-term use. Alternative medicine such as evening primrose oil (gamma-linolenic acid) was reported to increase submandibular and parotid salivary flow of patients with residual gland function (McCaul and Lamey, 1994).

Prednisolone and hydroxychloroquine, useful agents in rheumatoid arthritis, showed equivocal results in SSc patients with minimal clinical benefit (Pankhurst et al., 1996). However, this treatment produces immuno-modulation with a subsequent fall in the immune markers of disease such as a reduction in circulating auto-antibodies, erythrocyte sedimentary rate and excess production of immunoglobulins.

Chapter 6 Conclusion and Recommendations

This study concludes that

1. Tooth brushing twice daily was practised by Chinese people with Systemic Sclerosis in Hong Kong, and one-third of them performed regular interdental cleaning.
2. The caries experience of permanent teeth of Chinese people with Systemic Sclerosis in Hong Kong measured by DMFT was 11.4. They all had caries experience and 66% of them had active untreated decay.
3. None of the Chinese people with Systemic Sclerosis in Hong Kong were found periodontally healthy. Most of them (97%) had calculus found on their teeth and the majority (78%) required advanced periodontal treatment.
4. Over 80% of the Chinese people with Systemic Sclerosis in Hong Kong were found to have oral mucosal telangiectasia, but no other oral mucosal pathology was detected.
5. Many (71%) of the Chinese people with Systemic Sclerosis in Hong Kong experienced subjective dry-mouth symptoms.
6. Half of the Chinese people with Systemic Sclerosis in Hong Kong were found to have reduced salivary flow rate. However, the salivary pH were found to be comparable to that of the normal population.
7. About 20% of the Chinese people with Systemic Sclerosis in Hong Kong were not satisfied with their dental health.

From the findings of this study, we recommend that

1. It is essential to provide oral health education to Chinese people with Systemic Sclerosis and motivate them to seek dental care because the great majority of them have periodontal disease and untreated caries.

2. People with Systemic Sclerosis may have a more frequent regular follow up maintenance care and comprehensive preventive care because they have a higher risk of periodontal disease. It is also difficult for them to receive restorative procedures and dental prostheses due to limited mouth opening and reduced salivary flow.

References

1. Alexandridis C, White SC (1984). Periodontal ligament changes in patients with progressive Systemic Sclerosis. Oral Surg Oral Med Oral Pathol; 58:113-118.

2. Avouac J, Sordet C, Depinay C, Ardizonne M, Vacher-Lavenu MC, Sibilia J, Kahan A, Allanore Y (2006). Systemic Sclerosis-associated Sjögren's syndrome and relationship to the limited cutaneous subtype: results of a prospective study of sicca syndrome in 133 consecutive patients. Arthritis Rheum; 54:2243-2249.

3. Betram U (1967). Xerostomia: clinical aspects, pathology, and pathogenesis. Acta Odontol Scand; 25(suppl 49):15-21.

4. Chaffee NR (1998). CREST syndrome: clinical manifestations and dental management. J Prosthodont; 7:155-160.

5. Chang B, Schachina L, Whhite B, Wigkey FM, Wise RA (2006). Natural History of Mild–Moderate Pulmonary Hypertension and the Risk Factors for Severe Pulmonary Hypertension in Scleroderma. J Rheumatol; 33:269-274.

6. Cipoletti JF, Buckingham RB, Barnes EL, Peel RL, Mahmood K, Cignetti FE, Pierce JM, Rabin BS, Rodnan GP (1977). Sjogren's syndrome in progressive Systemic Sclerosis. Ann Intern Med; 87:535-541.

7. Department of Health HKSAR (2002). Oral Health Survey 2001

8. Drosos AA, Andonopoulos AP, Costopoulos JS, Stavropoulos ED, Papadimitriou CS, Moutsopoulos HM (1988). Sjogren's syndrome in progressive Systemic Sclerosis. J Rheumatol;15:965-968.

9. Eversole LR, Jacobsen PL, Stone CE (1984). Oral and gingival changes in Systemic Sclerosis (scleroderma). J Periodontol; 55:175-178.

10. Fox PC, Atkinson JC, Macynski AA, Wolff A, Kung DS, Valdez IH, Jackson W, Delapenha RA, Shiroky J, Baum BJ (1991). Pilocarpine treatment of salivary gland hypofunction and dry mouth (xerostomia). Arch Intern Med; 151:1149-1152.

11. Fox PC, Busch KA, Baum BJ (1987). Subjective reports of xerostomia and objective measures of salivary gland performance. J Am Dent Assoc;115:581-584.

12. Green D (1962). Scleroderma and its oral manifestation report of three cases of progressive Systemic Sclerosis (diffuse scleroderma). Oral Surg Oral Med Oral Pathol; 15:1312-1342.

13. Guern VL, Mahr A, Mouthon L, Jeanneret D, Carzon M, Guillevin L (2004). Prevalence of Systemic Sclerosis in a French multi-thnic country. Rheumatology (Oxford); 43:1129-1137.

14. Helmick CG, Felson DT, Lawrence RC, Gabriel S, Hirsch R, Kwoh CK, Liang MH, Kremers HM, Mayes MD, Merkel PA, Pillemer SR, Reveille JD, Stone JH; National Arthritis Data Workgroup (2008). Estimates of Prevalence of Arthritis and Other Rheumatic Conditions in the United States Part I. Arthritis Rheum; 58:15-25.

15. Hopper FE, Giles AD (1982). Orofacial changes in Systemic Sclerosis-report of a case of resorption of mandibular angles and zygomatic arches. Br J Oral Surg; 20: 129-134.

16. Kumar V, Cotran RS, Stanley L. Robins Basic Pathology. Philadelphia, PA: Saunders, 2003.

17. Landtwing K (1978). Evaluation of the normal range of vertical mandibular opening in children and adolescents with special reference to age and stature. J Maxillofac Surg; 6:157-162.

18. LeRoy EC, Black C, Fleischmajer R. (1988). Sclerderma (Systemic Sclerosis): classification, subset and pathogensis. J Rheumatol; 15:202-205.

19. Lin AT, Clements PJ, Furst DE (2003) Update on disease-modifying antirheumatic drugs in the treatment of Systemic Sclerosis. Rheum Dis Clin North Am; 29:409-426.

20. Leung KC, McMillan AS, Wong MC, Leung WK, Mok MY, Lau CS (2008). The efficacy of cevimeline hydrochloride in the treatment of xerostomia in Sjögren's syndrome in southern Chinese patients: a randomised double-blind, placebo-controlled crossover study. Clin Rheumatol; 27:429-436.

21. Magnant J, de Monte M, Guilmot JL, Lasfargues G, Diot P, Asquier E, Degenne D, Boissinot E, Diot E (2005). Relationship between occupational risk factors and severity markers of Systemic Sclerosis. J Rheumatol; 32:1713-1718.

22. McCaul JA, Lamey PJ (1994). Multiple oral mucoceles treated with gamma-linolenic acid: report of a case. Br J Oral Maxillofac Surg; 32:392-393.

23. Maître A, Hours M, Bonneterre V, Arnaud J, Arslan MT, Carpentier P, Bergeret A, de Gaudemaris R (2004). Systemic Sclerosis and occupational risk factors: role of solvents and cleaning products. J Rheumatol; 31:2395-2401.

24. Mayes MD (2003). Scleroderma epidemiology. Rheum Dis Clin N Am; 29:239-254.

25. McCaul JA, Lamey PJ (1994). Multiple oral mucoceles treated with gamma-linolenic acid: report of a case. Br J Oral Maxillofac Surg; 32:392-393.

26. McMillan AS, Leung KC, Leung WK, Wong MC, Lau CS, Mok TM (2004a). Impact of Sjögren's syndrome on oral health-related quality of life in southern Chinese. J Oral Rehabil; 31:653-659.

27. McMillan AS, Pow EH, Leung WK, Wong MC, Kwong DL (2004b). Oral health-related quality of life in southern Chinese following radiotherapy for nasopharyngeal carcinoma. J Oral Rehabil; 31:600-608.

28. McMillan AS, Tsang CS, Wong MC, Kam AY (2006). Efficacy of a novel lubricating system in the management of radiotherapy-related xerostomia. Oral Oncol; 42:842-848.

29. Mezitis M, Rallis G, Zachariades N (1989). The normal range of mouth opening. J Oral Maxillofac Surg; 47:1028-1029.

30. Nagy G, Kovács J, Zeher M, Czirják L (1994). Analysis of the oral manifestations of Systemic Sclerosis. Oral Surg Oral Med Oral Pathol; 77:141-146.

31. Navazesh M ,Christensen CM (1982). A comparison of whole mouth resting and stimulated salivary measurement procedures. J Dent Res; 61:1158-1162.

32. Naylor WP (1982). Oral management of the scleroderma patient. J Am Dent Assoc; 105:814-817.

33. Naylor WP, Douglass CW, Mix E (1984). The nonsurgical treatment of microstomia in scleroderma: a pilot study. Oral Surg Oral Med Oral Pathol; 57:508-511.

34. Osial TA Jr, Whiteside TL, Buckingham RB, Singh G, Barnes EL, Pierce JM, Rodnan GP (1983). Clinical and serologic study of Sjogren's syndrome in patients with progressive Systemic Sclerosis. Arthritis Rheum; 26:500-508.

35. Pankhurst CL, Smith EC, Rogers JO, Dunne SM, Jackson SH, Proctor G (1996). Diagnosis and management of the dry mouth: Part 1. Dent Update; 23:56-62.

36. Pizzo G, Scardina GA, Messina P (2003). Effects of a nonsurgical exercise program on the decreased mouth opening in patients with systemic scleroderma. Clin Oral Investig; 7:175-178.

37. Poole JL, Brewer C, Rossie K, Good CC, Conte C, Steen V (2005). Factors related to oral hygiene in persons with scleroderma. Int J Dent Hyg; 3:13-17.

38. Rasker JJ, Jayson MI, Jones DE, Matthews R, Burton JL, Rhys Davies E, Burton PA (1990). Sjogren's syndrome in Systemic Sclerosis: a clinical study of 26 patients. Sacnd J Rheumatol; 19: 57-65.

39. Raynaud's & Scleroderma Association. Oral and Dental Aspects of Scleroderma. Cheshire: Raynaud's & Scleroderma Association, 2008.

40. Rodan GP, Benedek TG. (1962). An Historical Account of the Study of Progressive Systemic Scelerosis (Diffuse Sceleroderma). Ann Intern Med; 57:305-318.

41. Rout PG, Hamburger J, Potts AJ (1996). Orofacial radiological manifestations of Systemic Sclerosis. Dentomaxillofac Radiol; 25:193-196.

42. Samet N, Tau S, Findler M, Susarla SM, Findler M (2007). Flexible, removable partial denture for a patient with Systemic Sclerosis (scleroderma) and microstomia: a clinical report and a three-year follow up. Gent Dent; 55:548-551.

43. Scardina GA, Messina P (2004). Systemic Sclerosis: description and diagnostic role of the oral phenomena. Gen Dent; 52:42-47.

44. Scardina GA, Pizzigatti ME, Messina P (2005). Periodontal Microciculatory Abnormaolities in Patients with Systemic Sclerosis. J Periodontol; 76:1991-1995.

45. Scully C, Cawson RA. Medical Problems in Dentistry. New York: Churchill Livingstone, 1998.

46. Scussel-Lonzetti L, Joyal F, Raynauld JP, Roussin A, Rich E, Goulet JR, Raymond Y, Senécal JL (2002). Predicting mortality in Systemic Sclerosis: analysis of a cohort of 309 French Canadian patients with emphasis on features at diagnosis as predictive factors for survival. Medicine (Baltimore); 81:154-167.

47. Sheppard IM, Sheppard SM (1965). Range of condylar movement during mandibular opening. J Prosthet Dent;15 :263-71.

48. Thomson WM (2007). Measuring change in dry-mouth symptoms over time using the Xerostomia Inventory. Gerodontology; 24:30-35.

49. Thomson WM, Chalmers JM, Spencer AJ, Williams SM (1999). The Xerostomia Inventory: a multi-item approach to measuring dry mouth. Community Dent Health; 16:12-17.

50. Thomson WM, Williams SM (2000). Further testing of the Xerostomia Inventory. Oral Surg Oral Med Oral Pathol Oral Radiol Endod; 89:46-50.

51. US Department of Health and Human Service National Institutes of Health National Institutes of Arthritis and musculoskeletal and skin disorder (2007). Handbook on Health – Scleroderma.

52. Waldrop RW, Heggie AAC (1987). Progressive Systemic Sclerosis –Orofacial manifestations case report. Aust Dent J; 32:258-262.

53. Wood GD, Branco JA (1979). A comparison of three methods of measuring maximal opening of the mouth. J Oral Surg; 37:175-177.

54. Wood GD, Branco JA.Wood RE, Lee P (1988). Analysis of the oral manifestations of Systemic Sclerosis (scleroderma). Oral Surg Oral Med Oral Pathol; 65:172-178.

55. Yenisey M, Külünk T, Kurt S, Ural C (2005). A prosthodontic management alternative for scleroderma patients. J Oral Rehabil; 32:696-700.

Appendix 1 –Progress note

Date	Event
30 Nov 2007	Brain storming of possible community health project
7 Dec 2007	Finalizing the project title
14 Dec 2007	Literature review and preparation of project protocol
25 Jan 2008	Initial contact with Rheumatology Clinic, Queen Mary
28 Jan 2008	Confirmation of project topic and finalizing project proposal
30 Jan 2008	Preparation of materials and instrument
1 Feb 2008	Trial on salivary function test
4 Feb 2008	Clinical photo taking exercise
5 Feb 2008	Calibration exercise on clinical diagnosis
13 – 15 Feb 2008	Telephone invitation of project participants
22 Feb 2008	Project rehearsal
25 – 29 Feb 2008	Main study at Prince Philip Dental Hospital
5 Mar 2008	Evaluation of the project
7 Mar 2008	Data entry completed
21 Mar 2008	Data analysis and report writing
23 May 2008	Submission of community health report
16 Jun 2008	Oral presentation of the community health project

Appendix 2- Consent (Chinese version)

敬啓者：

香港硬皮症病人口腔健康情況

硬皮症是一種影響皮膚及內臟組織的疾病。研究報告顯示，硬皮症病人患有蛀牙，牙齒侵蝕，張口困難和口乾的情況比較普遍，而患上黏膜病症(如念珠菌感染)及牙周病的風險亦較高。可是目前相關中國硬皮症病人口腔狀況的研究報告卻相當缺乏。

有見及此，香港大學牙醫學院現正在進行一項有關香港硬皮症病人口腔特徵及健康情況的專題研究。現誠邀 閣下參與是項研究。研究項目將包括填寫一份簡單的問卷,口腔檢查及唾液功能測試。檢查結果將會記錄於一份簡短的檢查報告上，以備參考。

閣下提供的所有資料將絕對保密及祇供研究用途，資料亦會於研究結束後毀掉。閣下可選擇不參與是項研究或隨時在不提供原因的情況下退出。 閣下於是次研究作出的任何決定不會影響 閣下接受醫療服務的權利。如有疑問，請致電 2859 0287 與朱振雄醫生聯絡。

多謝合作！

此致
研究參與者

香港大學牙醫學院
副教授 朱振雄醫生

回條

本人, _____, 香港身份證號碼 _____ , 明白這項研究的目的及內容並同意參與上述研究。

日期: _____ 簽署: _____

Appendix 3- Consent (English version)

Dear Sir/Madam,

Oral health status and oral features of Chinese people with Systemic Sclerosis in Hong Kong

Systemic scleroderma is a group of diseases that involves abnormal growth of connective tissue which supports the skin and internal organs. Case reports show that people with SS have high prevalence of dental caries, teeth erosion, small mouth (microstomia) and dry mouth (xerostomia), also an increased risk of gum (periodontal) infection and mucosal disease (candidal infection). However, there is no study on Chinese population.

The Faculty of Dentistry of the University of Hong Kong is going to conduct a study on systemic scleroderma patients. The aim is to examine the oral health status and features of Chinese people with systemic scleroderma. We would like to invite you to participate in this study. We will ask you to complete a questionnaire, perform a salivary function test followed by a clinical examination. An oral examination report will be given to you for your reference.

All information collected will be kept confidential and used for research purpose, it will be destroyed when the study is completed. You can choose not to participate in this study and you may withdraw anytime during the study period without giving any reasons. This will not affect your right in obtaining health care services. If you have any questions, you may contact Dr C H Chu at 2859 0287. Please complete the following consent if you agree to join our study.

Thank you.

Dr CH Chu
Associate Professor
Faculty of Dentistry
The University of Hong Kong

Reply Slip

I, _____, I/D No. _____ understand the aim and content of the study and agree to participate in this study.

Date: _____ Signature: _____

Appendix 4 – Photo taking consent form

PHOTOGRAPH RELEASE

I, _____
(Patient's Name) (Hospital Registration No.)

do hereby consent to and authorize The Prince Philip Dental Hospital and the Faculty of Dentistry, University of Hong Kong to use all or any photographs taken of me, regardless of modifications or alterations, for the purposes of education, research records and /or publication.

Signature _____

Address _____

Date _____

If the person is a minor:–

I the undersigned, hereby warrant that I am the parent or guardian of _____
(Patient's Name)

_____ and having read the foregoing, do hereby consent to those matters
(Registration No.)

stated above.

Signature _____

Address _____

Date _____

採用照片之同意書

本人 _____
（病人姓名） （登記號碼）

茲同意及授權菲臘牙科醫院及香港大學牙醫學院採用爲本人所拍攝之所有或部份照片，不論有無經過修改或更改，作爲教育，研究或發表用途。

簽名 _____

地址 _____

日期 _____

未成年病人：_____

本人茲證明爲 _____
（病人姓名） （登記號碼）

之家長／監護人。現經細讀上述之要求，特表同意。

簽名 _____

地址 _____

日期 _____

Appendix 5 – Medical history record form

THE PRINCE PHILIP DENTAL HOSPITAL

Name of Patient .....

MEDICAL HISTORY
病歷

1.	Y	N	Poor General Health	健康不良
2.	Y	N	Current Medical Treatment	是否正在接受醫藥治療
3.	Y	N	Medicine Pills or Tablets	是否服食任何藥物
4.	Y	N	Previous Hospital Experience	入住醫院或在醫院「門診部」接受治療
5.	Y	N	General Anaesthetic	「全身痲醉」
6.	Y	N	Allergic to Penicillin or Other Substances	對「盤尼西林」、食物或其他藥物有「敏感」
7.	Y	N	Hay Fever, Eczema, Asthma	「乾草熱」、「濕疹」或「哮喘」
8.	Y	N	Rheumatic Fever	「風濕性熱病」
9.	Y	N	Abnormal Bleeding	施手術、脫牙或受傷時是否流血不止
10.	Y	N	Steroid, Anti-coagulant or Irradiation Therapy	「類固醇」、「抗凝血藥物」或「電療」
11.	Y	N	Jaundice, Hepatitis or Other Liver Disease	「黃膽病」、「肝炎」或其他「肝病」
12.	Y	N	Fainting or Giddiness	突然暈倒或常覺頭暈
13.	Y	N	Childhood Diseases	曾否患「水痘」、「痲疹」、「風疹」、「疼腮」等症
14.	Y	N	Expectant Mother	是否懷孕

Heart Disease	心臟病	( )	Kidney Disease	腎病	( )	T.B.	結核病(肺癆)	( )
Hypertension	高血壓	( )	Diabetes	糖尿病	( )	Epilepsy	羊癇	( )
Blood Disease	血病	( )	Thyroid Disease	甲狀腺病	( )	Stroke	腦充血	( )
V.D.	性病	( )	Other Medical Problems	其他有關健康資料	.....			

Date ..../..../.... Completed By : ..... No : .....

DRUGS

.....

.....

.....

NOTES

.....

.....

.....

.....

.....

.....

.....

Appendix 6 – Questionnaire (Chinese version)

姓名: _____ 年齡 _____ 性別 男 / 女

請圈出適當的答案。

Q1 你上一次見牙醫是什麼時候？

1. 一年之內 2. 一年之前 三年之內 3. 三年以前/從來沒有

Q2 你多久刷一次牙?

1. 一日一次 2. 一日兩次以上 3. 間中

Q3 你上次何時用過牙簽清潔牙縫?

1. 昨天 / 今天 2. 一星期內 3. 從來不用

Q4 你上次何時用過牙線或牙縫刷清潔牙縫？

1. 昨天 / 今天 2. 一星期內 3. 從來不用

Q5 你上次何時用過漱口水？

1. 昨天 / 今天 2. 一星期內 3. 從來不用

Q6 你覺得現時自己有蛀牙和牙周病嗎?

6a 蛀牙 1. 有 2. 沒有(請到第 9 題) 3. 不知道

6b 牙周病 1. 有 2. 沒有(請到第 9 題) 3. 不知道

Q7 你有去找牙醫處理問題嗎?

1. 有 (請到第 9 題) 2. 沒有

Q8 爲甚麼沒有去找牙醫處理問題?

1. 怕見牙醫 2. 怕痛 3. 收費太高

4. 沒有時間 5. 到診所有困難 6. 其他

Q9 你滿意你現時的牙齒健康狀況嗎?

1. 不滿意 2. 可以接受 3. 滿意

Q10 你現在或過去有沒有吸煙的習慣？

1. 有, 如有，Q10a 煙齡多少年？ _____ 2. 無

Q11 你現在或過去有沒有飲烈酒的習慣，(例如威士忌、拔蘭地、米酒等)？

1. 有, 如有，Q11a 你已飲了幾多年？ _____ 2. 無

全問卷完 謝謝!

Appendix 7 – Questionnaire (English version)

Name: _____ **Age** _____ **Sex** **M/ F**

Please circle the most appropriate answer.

Q1 When did you last see a dentist?

1. Within a year 2. More than a year, less than 3 years 3. More than 3 years / never

Q2 How often do you brush your teeth?

1. Once daily 2. Twice or more daily 3. Occasionally

Q3 When did you last clean the area between teeth with toothpick?

1. Today / yesterday 2. Within a week 3. More than a week/ Never

Q4 **When did you last clean the area between teeth with floss or brush?**

1. Today / yesterday 2. Within a week 3. More than a week/ Never

Q5 When did you last use mouthrinse?

1. Today / yesterday 2. Within a week 3. More than a week/ Never

Q6 Do you think you have tooth decay and periodontal problem?

6a Tooth decay 1. Yes 2. No (To Q9) 3. Don't know

6b Periodontal problem 1. Yes 2. No (To Q9) 3. Don't know

Q7 Did you seek for treatment from a dentist?

1. Yes (To Q9) 2. No

Q8 Why didn't you seek treatment from a dentist?

1. Afraid of dentist 2. Afraid of pain 3. High dental fee

4. No Time 5. Difficult to go to a dental clinic 6. Other reason : _____

Q9 Are you satisfied with you dental health?

1. No 2. Acceptable 3. Yes

Q10 Are you a smoker ?

1. Yes. Q10a Smoking experience: _____ years 2. No

Q11 Are you an alcoholic drinker?(example: Whiskey, Brandy, Wheat wine)

1. Yes. Q11a Drinking experience: _____ years 2. No

The End. Thank you!

Appendix 8 – Xerostomia Inventory (Chinese version)

口乾症問卷

Xerostomia Inventory

Name: _____ Date: _____

PPDH no.: _____ Group: _____

你有幾多時候

		從未試過	好少	間中	多數	通常
1.	要飲住水才能把食物吞下	☐1	☐2	☐3	☐4	☐5
2.	吃東西時會覺得口乾	☐1	☐2	☐3	☐4	☐5
3.	覺得口唇好乾	☐1	☐2	☐3	☐4	☐5
4.	覺得有些食物是很難吞的	☐1	☐2	☐3	☐4	☐5
5.	覺得口乾	☐1	☐2	☐3	☐4	☐5
6.	需要半夜起床飲水	☐1	☐2	☐3	☐4	☐5
7.	覺得眼睛好乾	☐1	☐2	☐3	☐4	☐5
8.	覺得食乾的食物是很困難的	☐1	☐2	☐3	☐4	☐5
9.	覺得鼻入面也很乾	☐1	☐2	☐3	☐4	☐5
10.	要啜糖或者酸的東西來舒緩口乾	☐1	☐2	☐3	☐4	☐5
11.	覺得面部皮膚好乾	☐1	☐2	☐3	☐4	☐5

Appendix 9 – Xerostomia Inventory (English version)

Xerostomia Inventory

Please answer the following questions by checking the box that best represents your answer.

		Never	Hardly ever	Occasionally	Fairly often	Very often
1.	I sip liquids to aid in swallowing food	☐1	☐2	☐3	☐4	☐5
2.	My mouth feels dry when eating a meal.	☐1	☐2	☐3	☐4	☐5
3.	My lips feel dry.	☐1	☐2	☐3	☐4	☐5
4.	I have difficulties swallowing certain foods.	☐1	☐2	☐3	☐4	☐5
5.	My mouth feels dry.	☐1	☐2	☐3	☐4	☐5
6.	I get up at night to drink.	☐1	☐2	☐3	☐4	☐5
7.	My eyes feel dry.	☐1	☐2	☐3	☐4	☐5
8.	I have difficulty in eating dry foods.	☐1	☐2	☐3	☐4	☐5
9.	The inside of my nose feels dry.	☐1	☐2	☐3	☐4	☐5
10.	I suck sweets or cough lollies to relieve dry mouth.	☐1	☐2	☐3	☐4	☐5
11.	The skin of my face feels dry.	☐1	☐2	☐3	☐4	☐5

Appendix 10 – Charting record form

Name: _____ Age _____ Sex: M / F

Examiner: Mandy / Lillian

Dentition

18	17	16	15	14	13	12	11	21	22	23	24	25	26	27	28
48	47	46	45	44	43	42	41	31	32	33	34	35	36	37	38

0	Sound	5	Missing, any other reason
1	Decayed	6	Fissure sealant
2	Filled, with decay	7	Bridge abutment/crown/veneer/implant
3	Filled, without decay	8	Unerupted, unexposed
4	Missing, as a result of caries	9	Not recorded

Oral Mucosa Assessment

Condition	Location

Condition		Location	
1	No abnormal condition	1	Vermillion border
2	Malignant tumour	2	Commissures
3	Leukoplakia	3	Lips
4	Ulceration	4	Buccal mucosa
5	Acute Necrotizing gingivitis	5	Floor of Mouth
6	Candidiasis	6	Tongue
7	Abscess	7	Hard and/or soft palate
8	Others _____	8	Alveolar ridge / gingiva
9	Not recorded	9	Not recorded

Mouth Opening

Inter-incisal Distance		

	Telangiec-tasia	Upper Denture	Lower Denture	Tongue Tie	TMJ	
					Clicking	Tenderness
1 / 0						

CPI			
			0 = Healthy 1 = Bleeding after probing 2 = Calculus, all black band visible 3 = Pocket 4 – 5 mm, gingival margin within black band 4 = Pocket >6mm, black band not visible X = Excluded sextant (<2 teeth present) 9 = Not recorded

Appendix 11 – Oral examination report (Chinese version)

香港大學牙醫學院
硬皮症病人口腔情況調查

此致 ： _____ 先生/小姐

口腔檢查撮要報告

多謝　閣下同意參加於菲臘牙科醫院內舉行的口腔特徵及情況調查。這是一個口腔檢查及唾液功能測試。

在檢查過程中我們發現　閣下最少有_____隻蛀牙，唾液腺分泌爲每分鐘 _____毫升(較正常爲少 / 正常 / 較正常爲多)，
酸鹼值(pH)爲 _____ (較正常爲低 / 正常 / 較正常爲高低)。

閣下的口腔情況 良好 / 一般 / 欠佳。

我們建議　閣下要加強口腔護理及 在短期內 / 定期 去看牙醫作口腔檢查。

閣下刷牙時要特別注意清潔下列牙齒 :

☐上顎門牙
☐上顎大牙
☐下顎門牙
☐下顎大牙

Appendix 12 – Oral examination report (English version)

THE UNIVERSITY OF HONG KONG
Faculty of Dentistry

Study on oral health status and oral features of Chinese people with Systemic Sclerosis

Dear Mr. /Ms_____,

Oral Examination Report

Thank you for your participation in the study of oral health status and oral features of Chinese people with systemic sclerosis which was held in the Prince Philip Dental Hospital. This consisted of an oral examination and a salivary function test.

In our clinical examination, it was found that you have at least _____ decayed tooth / teeth, your salivary secretion of _____ml/min (which is less than normal / normal / more than normal) and your salivary pH value is of _____ (which is lower than normal / normal / higher than normal).

Your oral hygiene is rated as fair / average / satisfactory.

We would suggest you to reinforce oral health care and also visit a dentist immediately / regularly for dental check-up.

Please pay particular attention in cleaning the following teeth when you perform daily brushing listed as below:

☐ Upper front teeth
☐ Upper back teeth
☐ Lower front teeth
☐ Lower back teeth